

Venous Malformations of the Oral Cavity in Children: A Review Article

Mungnirandr A, MD¹

¹ Division of Pediatric Surgery, Department of Surgery, Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok, Thailand

Venous malformations of the oral cavity present with various clinical pictures. Since venous malformation is not a vascular tumor, spontaneous resolution of the lesion should not be expected. There are many modalities for treatment of venous malformation of the oral cavity, and these modalities are herein reviewed.

Keywords: Venous malformation, Oral cavity, Pediatric

J Med Assoc Thai 2020;103(Suppl.2): 109-11

Website: <http://www.jmatonline.com>

Venous malformations of the oral cavity present with various clinical pictures. Some localize on the superficial surfaces as venous lake, and others infiltrate deeper structures and may extend to structures beyond the oral cavity. Since venous malformation is not a vascular tumor, such as hemangioma, spontaneous resolution of the lesion should not be expected. However, spontaneous enlargement of venous malformation during puberty and menopause has been reported⁽¹⁾. There are many modalities for treatment of venous malformation of the oral cavity, and these modalities are herein reviewed.

Conservative treatment is often the first-line approach in asymptomatic patients. Treatments include routine elastic bandage or stockinette application. The aim is to collapse superficial venous malformation to prevent venous stasis, thrombus formation, and engorged vein. Non-steroidal anti-inflammatory drugs (NSAIDs) are used for analgesia in venous malformation with thrombophlebitis. In thrombophlebitic patients with high D-dimer level, low molecular weight heparin is recommended⁽²⁾.

Sclerotherapy is used to induce injury to vascular malformation, which results in the subsequent development of thrombosis. Many different agents can be used, including alcohol, bleomycin, and pingyangmycin. Alcohol reduced the size and color of venous malformation in 84 to 100% cases, and 27 to 68% of patients had complete resolution. Complications developed in 18% of children and the majority involved nerve and skin⁽³⁾. Hemoglobinuria and cardiac arrhythmia have also been reported⁽⁴⁾. Temporary tracheotomy to facilitate direct intralesional ethanol

sclerotherapy of extensive venous malformations with oropharyngeal involvement was reported to be a safe and effective method⁽⁵⁾. Pingyangmycin is a new agent that is used for treatment of venous malformation with reported 95% partial resolution, 46 to 100% complete resolution, and 2% minor skin and subcutaneous tissue complications⁽³⁾. Different concentrations of pingyangmycin for venous malformation at different sites can reduce complications⁽⁶⁾. A unique case of tongue squamous cell carcinoma after pingyangmycin treatment was reported⁽⁷⁾. Foam sclerotherapy technique demonstrated increased effectiveness by



Figure 1. Before laser and sclerotherapy treatment (A, C). After laser and sclerotherapy treatment (B, D).

Correspondence to:

Mungnirandr A.

Division of Pediatric Surgery, Department of Surgery, Faculty of Medicine Siriraj Hospital, Mahidol University, 2 Wanglang Road, Bangkoknoi, Bangkok 10700, Thailand.

Phone: +66-2-4198027, **Fax:** +66-2-4129160

E-mail: akkrapol.mun@mahidol.ac.th

How to cite this article: Mungnirandr A. Venous Malformations of the Oral Cavity in Children: A Review Article. J Med Assoc Thai 2020;103(Suppl.2): 109-11.

decreasing the rate of sclerosing agent elimination, and by increasing the amount of time the sclerosing agent is in contact with the endothelium⁽⁸⁾. However, foam may overflow into a nearby structure and cause injury or necrosis. Radio-opaque foam guided by digital subtraction angiography technique can reduce complications by reducing the risk of infiltration of sclerosing agent into surrounding tissues or adjacent vessels⁽⁹⁾.

Some types of lasers cause selective damage to hemoglobin, which results in damage to the endothelium of venous malformation. Laser therapy can be applied via transcutaneous technique for treatment of superficial lesions, and by way of interstitial or endovascular technique for treatment of deeper lesions. Transcutaneous laser coagulation is used for superficial venous malformation^(10,11). Various parameters, such as shrinkage of lesion or disappearance of color, were proposed as the endpoints of treatment⁽¹²⁾. Nd-YAG laser has a long wavelength (1,064 nm) and is supposed to penetrate deeper into venous malformation^(10,12). Thermal trauma can be prevented or minimized by using underwater or ice technique^(10,13).

Interstitial and endovascular laser coagulation is used for deeper venous malformation. The laser beam is produced by a fiber-optic beam delivery system^(14,15). Thermal injury into the venous malformation was suspected based on observable injury to the overlying normal tissue. Improved accuracy of the localization of venous malformation can be obtained by Doppler ultrasonography. In large venous malformation, compartmentalization can be used to limit the irradiated area to avoid severe reactions and excessive thermal injury to nearby tissues and structures⁽¹⁶⁾. Endovenous laser coagulation can be used in children with minimal side effects⁽¹⁷⁾; however, other associated vascular anomalies and venous insufficiency are commonly found⁽¹⁸⁾.

Surgery can be safely performed in small venous malformation; however, bleeding and cosmetic deformity are major complications of surgical excision. Bleeding from venous malformation can be ameliorated by using N-butyl-2-cyanoacrylate and pre-operative glue embolization^(19,20). Because venous malformation of lip result in tissue hypertrophy, 70% of the lip can be surgically excised without cosmetic deformities⁽²¹⁾.

Liquid nitrogen cryotherapy is an alternative treatment modality. Complete remission of lip mucous membrane venous malformation was reported in 77.4% of patients that did not develop complications. Radiofrequency can treat venous malformation by inducing interstitial thermal ablation. Smaller lesions were reported after adjunct treatment with other modalities, including surgical excision and/or bleomycin injection^(22,23). Excellent outcome without skin necrosis or nerve damage was reported in 62.9% of patients after electrochemical therapy combined with pingyangmycin⁽²⁴⁾.

Venous malformation is a subtype of vascular malformation. Venous malformations of the oral cavity have clinical manifestations that may be different from those



Figure 2. Transcutaneous laser coagulation.



Figure 3. Interstitial laser coagulation.



Figure 4. Venous malformation with phlebolith.

observed at other locations. Many modalities of treatment are proposed. Interdisciplinary treatments are required for large and/or complex venous malformations.

What is already known on this?

Already known: venous malformation is a congenital vascular malformation. Multimodality treatments are required for the best treatment.

What this study adds?

This review article added the new modality of knowledge: new treatment by laser and surgical technique. The use of new agent: pingyangmycin and its new complication: malignancy change.

Funding disclosure

This was an unfunded study.

Potential conflicts of interest

The author declares no conflicts of interest.

References

1. Maclellan RA, Goss JA, Greene AK. Vascular malformation enlargement during menopause. *J Craniofac Surg* 2018;29:1271-2.
2. Domp Martin A, Vikkula M, Boon LM. Venous malformation: update on aetiopathogenesis, diagnosis and management. *Phlebology* 2010;25:224-35.
3. Horbach SE, Lokhorst MM, Saeed P, de Gouyon Matignon de Pontouraude CM, Rothova A, van der Horst CM. Sclerotherapy for low-flow vascular malformations of the head and neck: A systematic review of sclerosing agents. *J Plast Reconstr Aesthet Surg* 2016;69:295-304.
4. Dasgupta R, Patel M. Venous malformations. *Semin Pediatr Surg* 2014;23:198-202.
5. Wang D, Su L, Han Y, Wang Z, Zheng L, Li J, et al. Direct intralesional ethanol sclerotherapy of extensive venous malformations with oropharyngeal involvement after a temporary tracheotomy in the head and neck: Initial results. *Head Neck* 2017;39:288-96.
6. Liu ZY, Xue L, Yuan WL, Wei JJ, Lei BC, Wang XK. The efficacy of treatment of oral and maxillofacial venous malformations by pingyangmycin injection with different concentrations. *Shanghai Kou Qiang Yi Xue* 2013;22:195-7.
7. Chen G, Cai X, Ren JG, Jia J, Zhao YF. Unexpected development of tongue squamous cell carcinoma after sclerotherapy for the venous malformation: a unique case report and literature review. *Diagn Pathol* 2013;8:182.
8. Ul HF, Mitchell SE, Tekes A, Weiss CR. Bleomycin foam treatment of venous malformations: A promising agent for effective treatment with minimal swelling. *J Vasc Interv Radiol* 2015;26:1484-93.
9. Chen AW, Liu YR, Li K, Zhang K, Wang T, Liu SH. Efficacy of sclerotherapy with radio-opaque foam guided by digital subtraction angiography for the treatment of complex venous malformations of the head and neck. *Br J Oral Maxillofac Surg* 2015;53:809-13.
10. Mungnirand A, Nuntasunti W, Manuskiatti W. Neodymium-doped yttrium aluminium garnet laser treatment of pediatric venous malformation in the oral cavity. *Dermatol Surg* 2016;42:875-9.
11. Frigerio A, Tan OT. Laser applications for benign oral lesions. *Lasers Surg Med* 2015;47:643-50.
12. John HE, Phen HS, Mahaffey PJ. Treatment of venous lesions of the lips and perioral area with a long-pulsed Nd:YAG laser. *Br J Oral Maxillofac Surg* 2016;54:376-8.
13. Crockett DJ, Meier JD, Wilson KF, Grimmer JF. Treatment of oral cavity venous malformations with the Nd:YAG laser using the underwater technique. *Otolaryngol Head Neck Surg* 2013;149:954-6.
14. Apfelberg DB. Intralesional laser photocoagulation-steroids as an adjunct to surgery for massive hemangiomas and vascular malformations. *Ann Plast Surg* 1995;35:144-8.
15. Alvarez-Camino JC, Espana-Tost AJ, Gay-Escoda C. Endoluminal sclerosis with diode laser in the treatment of orofacial venous malformations. *Med Oral Patol Oral Cir Bucal* 2013;18:e486-90.
16. Miyazaki H, Romeo U, Ohshiro T, Kudo T, Makiguchi T, Kawachi N, et al. Treatment strategies for large oral venous malformations using intralesional laser photocoagulation. *Lasers Med Sci* 2014;29:1987-90.
17. Patel PA, Barnacle AM, Stuart S, Amaral JG, John PR. Endovenous laser ablation therapy in children: applications and outcomes. *Pediatr Radiol* 2017;47:1353-63.
18. Mungnirand A, Zeeh U. Results of surgical repair of primary obstructive megaureter. *J Med Assoc Thai* 2006;89:329-33.
19. Idle MR, Monaghan AM, Lamin SM, Grant SW. N-butyl-2-cyanoacrylate (NBCA) tissue adhesive as a haemostatic agent in a venous malformation of the mandible. *Br J Oral Maxillofac Surg* 2013;51:565-7.
20. Tieu DD, Ghodke BV, Vo NJ, Perkins JA. Single-stage excision of localized head and neck venous malformations using preoperative glue embolization. *Otolaryngol Head Neck Surg* 2013;148:678-84.
21. De Castro DK, Ng ZY, Holzer PW, Waner M, Cetrulo CL Jr, Fay A. One-stage supramaximal full-thickness wedge resection of vascular lip anomalies. *J Oral Maxillofac Surg* 2017;75:2449-55.
22. Garg S, Kumar S, Singh YB. Intralesional radiofrequency in venous malformations. *Br J Oral Maxillofac Surg* 2015;53:213-6.
23. Civelek S, Sayin I, Ercan I, Cakir BO, Turgut S. Bipolar radiofrequency-induced interstitial thermoablation for oral cavity vascular malformations: Preliminary results in a series of 5 children. *Ear Nose Throat J* 2012;91:488-92.
24. Xue L, Cao RY, Xu DP, Sun NN, Tan HS, Wang XK. Percutaneous treatment of large venous malformations in the oral and maxillofacial regions using electrochemical therapy combined with pingyangmycin. *J Oral Maxillofac Surg* 2015;73:1384-91.

ทบทวนวรรณกรรม ความผิดปกติของหลอดเลือดดำของช่องปาก

อัครพล มุ่งรินทร์

ความผิดปกติของหลอดเลือดดำเป็นความผิดปกติแต่กำเนิดประเภทหนึ่ง ความผิดปกติของหลอดเลือดดำของช่องปากและริมฝีปากมีการรักษาได้หลายวิธี บทความนี้ได้ทบทวนวรรณกรรมสำหรับการรักษา ได้แก่ การรักษาแบบประคับประคอง การใช้แผ่นยางยืดรัด การฉีดยาเข้าในรอยโรค โดยการฝังรังสีวินิจฉัยร่วมรักษา การใช้ไนโตรเจนเหลว การใช้เลเซอร์คาร์บอนไดออกไซด์และเข้าหลอดเลือดดำ และการรักษาโดยวิธีการผ่าตัด การรักษาความผิดปกติของหลอดเลือดดำแต่กำเนิด โดยใช้สาขาวิชา จะได้รับผลการรักษาที่ดีที่สุด
